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A review of human sparganosis in Thailand

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Summary

Background: Sparganosis is a zoonosis that occurs occasionally in humans. The infection is reported in many countries but is most common in eastern Asia. In Thailand, a southeast Asian country, the infection is sporadic.
Design: In this study the clinical presentations of human sparganosis cases in Thailand were investigated by means of a literature review.
Results: Reports of 34 cases of sparganosis were found. The infections were ocular (17 cases), subcutaneous (ten cases), central nervous system (five cases), auricular (one case), pulmonary (one case), intraosseous (one case) and intraperitoneal (one case). Of these 34 cases, 14 had risk behaviour reported, 12 had a history of drinking impure water, five had a history of eating frog or snake meat and two had a history of using frog or snake meat as a poultice. Some cases had more than one risk factor.
Conclusion: Most cases of sparganosis in Thailand presented with superficial ocular mass lesions. The major risk behaviour in Thailand is drinking water contaminated with the infective organism. Some cases of serious deep visceral sparganosis have also been reported.

Introduction

Humans can serve as the intermediate host for several species of cestodes. For example, infections with *Taenia* spp. cause cysticercosis, and *Echinococcus* spp. cause hydatid disease.[1] Humans can also serve as the secondary intermediate host for some cestodes of the order Pseudophyllidea.[1] In these cases the metacestode stage, normally known as a plerocercoid, is called a sparganum, and the resulting infection is sparganosis.[1—4]

Sparganosis is a zoonosis contracted from amphibians, reptiles or mammals, which occurs occasionally in humans.[1,2,4] The infection is reported in many countries but is most common in eastern Asia.[1,4] In

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Thailand, a southeast Asian country, the infection is sporadic.[1,4] The first report of human sparganosis in Thailand was made by Daengsawang and Tansurat in 1943.[5] The patient presented with a swollen eyelid and underwent spontaneous removal of the parasite which was identified morphologically.[5] Since this report there have been sporadic reports of human sparganosis in Thailand. These are reviewed in the present study in order to present a clinical summary of sparganosis among Thai patients.

Materials and methods

This study was designed as a descriptive retrospective study. A literature review of the papers concerning sparganosis in Thailand was performed using the database of published works cited in Index Medicus and the Science Citation Index. The author also reviewed the published works in all 256 local Thai journals, which are not included in the international citation index, in the database ''Thai Index Medicus'' (161.200.96.233/thaiim.html). The literature review focused on the years 1943—2004. All relevant reports were obtained and the details of the clinical presentations of the patients in all included papers were studied.

Results

According to this study there have been 34 reported cases of sparganosis in Thailand since 1943 (Table 1).[6—27] The infections were ocular (17 cases),

Table 1 Reports of human sparganosis in Thailand since 1943.

Year	Authors	Age (years)	Sex	Organ	Risk habit **
1943	Tansurat[5]	39	Male	Eye	N/A
1950	Pradatsundrasar[6]	13	Female	Subcutaneous abdominal wall	N/A
1960	Sampavapon[7]	14	Female	Eye	A,B
1960	Sampavapon[7]	13	Female	Eye	A,B
1960	Samitalumpa[8]	11	Female	Eye	N/A
1964	Bedavanija and Namatra[9]	25	Female	Eye	C
1964	Jipipob and Chenpanich[10]	33	Female	Eye	A
1964	Tansurut[5]	19	Female	Subcutaneous abdominal wall	N/A
1965	Tansurut[5]	40	Male	Subcutaneous abdominal wall	N/A
1965	Tansurut[5]	18	Female	Subcutaneous forehead	N/A
1968	Tesacharoen[4]	N/A	Male	Eye	N/A
1968	Tesacharoen[4]	N/A	Female	Eye	N/A
1969	Jipiphob[11]	39	Male	Eye	C
1971	Pradatsundrasar et al.[12]	46	Female	Brain	N/A
1974	Khamboonraung et al.[13]	23	Female	Peritoneal cavity	A
1975	Tesacharoen[4]	24	Male	Subcutaneous neck	N/A
1978	Tesacharoen[4]	N/A	Male	Subcutaneous scrotum	N/A
1978	Tesacharoen[4]	17	Female	Subcutaneous groin region	N/A
1979	Tesacharoen[4]	23	Female	Subcutaneous thigh	N/A
1980	Tesacharoen[4]	60	Male	Subcutaneous thumb	N/A
1980	Masrungsan et al.[14]	34	Male	Eye	A,B
1981	Masrungsan et al.[15]	34	Female	Eye	N/A
1984	Prommakup and Chayaphai[16]	14	Male	Brain	N/A
1985	Pongprasert and Somboon[17]	25	Female	Brain	N/A
1988	Kittiponghansa et al.[18]	51	Male	Eye and ear	A
1989	Chaijukool and Ying-Yuad[19]	37	Male	Eye	N/A
1989	Jenchitr et al.[20]	37	Male	Eye	A
1989	Luckanakul[21]	26	Female	Eye	A,B
1992	Chayapum and Tungsinmunkong[22]	33	Male	Spinal cord and brain	N/A
1992	Kasantikul et al.[23]	51	Female	Eye	N/A
1993	Ausayakhun et al.[24]	30	Female	Eye	A
2000	Jirawattanasomkul and Noppakun[25]	32	Male	Subcutaneous many areas, brain	A
2001	Phunmanee et al.[26]	25	Male	Lung	A
2002	Settakorn et al.[27]	51	Male	Leg bone	A,B

*N/A means data not available; ** risk habit: A = drinking impure water, B = eating frog or snake meat, C = use of frog or snake meat poultice.

subcutaneous (ten cases), central nervous system (CNS) (five cases), auricular (one case), pulmonary (one case), intraosseous (one case) and intraperitoneal (one case) (Table 1). There was one case of combined ocular and auricular infection and another case of combined subcutaneous and CNS infection. Ocular sparganosis was the most common presentation, with most cases presenting as an extraocular mass. Subcutaneous infections were most common in the abdominal wall (three cases). CNS infections were all intracranial, with one patient having both brain and spinal infection.

The mean age was 34.1 ± 12.8 years (range 11–60 years) with a male:female ratio of 15:19. Risk behaviours were not recorded in most cases. Of the 34 cases, 14 had identified risk behaviour, 12 had a history of drinking impure water, five had a history of eating frog or snake meat and two had a history of using frog or snake meat as a poultice. Some cases had more than one risk behaviour. All cases presented with abnormal mass lesions by routine physical examination, surgical procedure or imaging and were diagnosed by identification of the worm morphologically or histologically. All cases, except for one lethal case with deep brain infection[31] made a complete recovery after removal of the worm.

Discussion

The term sparganosis is defined as an infection by the larvae of parasitic tapeworms of *Spirometra* species.[1–4] Humans are an intermediate host for the parasite and infection occurs in several ways including (a) drinking water containing infected copepods, which are small crustaceans, (b) eating amphibian, reptile or rodent meat and (c) using those meats as a poultice; a traditional self-treatment according to ritual belief.[1,4,28] These risk behaviours were identified in 41.2% of cases.

According to a recent study by Park et al., a history of eating raw snake or frog is the most important risk factor for sparganosis with an odds ratio of 3.1.[29] However, eating raw meat is not the most common identified risk habit in the present study. The drinking of impure water, which can be a direct contact to the first intermediate host, was the most common risk behaviour as it was also in the report by Norman and Kreutner.[30] Indeed, eating raw frog or snake meat is not a traditional eating habit in Thailand. Although some ethnic populations in rural areas eat this meat, it is eaten cooked. The problem of impure water can be seen in remote rural areas where running water is not available. Although this problem has been seen in the USA in the past,[31] it still occurs in Thailand today, with several cases during previous decades citing the risk behaviour of drinking impure water.

Once ingested by a human the spargana larvae undergo visceral migration and can end up in many tissues, where they grow. The lengths of the larvae recorded in Thailand were up to 2 mm.[4] Ocular sparganosis was the most common presentation in this review. This superficial infection is also common in other countries.[30,32,33] Mass lesion was the most common clinical sign, as can be seen in other reports.[30] Some serious presentations, especially deep visceral infections, were seen in this review. CNS sparganosis cases usually have the worst prognosis. These cases presented with the symptom of repeated headache and were finally diagnosed by intracranial surgery. As in the study of Holodniy et al., progressive neurodeficit was seen in CNS sparganosis in Thailand.[34] A case with combined sparganosis and cysticercosis was also seen.[17]

The rarest presentations of sparganosis, including pulmonary, intraosseous and intraperitoneal were seen in this series. The patient with pulmonary sparganosis first presented with pulmonary symptoms; a chest X-ray revealed diffuse multiple masses with cavitations, while bronchoscopy with a transbronchial lung biopsy was non-diagnostic.[17] Indeed, there are some reports of pulmonary lesions, described as pulmonary proliferating,[35–49] due to intrapulmonary migration of sparganum but finding the worm in the lungs is very rare. With respect to intraosseous sparganosis, the Thai case included here is the first ever reported.[27] In this case, the patient primarily complained of a slow-growing painful mass on the right leg. The radiographic findings showed an infiltrative osteolytic lesion with

Table 2 Summary of presentations and risk habits of Thai patients with human sparganosis.

Presentations and risk factor	Percentage[a]
Presentations	
• ocular	50.0
• subcutaneous	29.4
• central nervous system	14.7
• auricular	2.9
• pulmonary	2.9
• intraosseous	2.9
• intraperitoneal	2.9
Risk habits	
• drinking impure water	35.3
• ingestion of frog or snake meat	14.7
• use frog or snake meat poultice	5.9
• unknown	58.8

[a] Some cases have more than one presentation and some have more than one risk factor.

speckled calcification at the proximal tibia, the clinical diagnosis of which favoured bone cancer. The Thai case of intraperitoneal sparganosis is also the first ever reported.[13] The patient had abdominal pain and discomfort as well as an abdominal mass. Some additional cases of intraperitoneal sparganosis have been reported in Japan[40] and the USA.[41] For deep sparganosis infections, additional therapy with albendazole is recommended.[41—42]

An important limitation of this study should be mentioned. Because this is a collection of published case reports there will be a publication bias towards rare complications of the disease rather than the most common clinical manifestations. In conclusion, most sparganosis cases reported from Thailand presented with superficial ocular mass lesions. The major risk behaviour in Thailand is drinking contaminated water (Table 2). Some cases of serious deep visceral sparganosis have also been reported. Surgical excision of the parasite resulted in a cure of disease in most cases.

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